

Title:

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Recurrent hemobilia secondary to extrahepatic biliary tract cholangiocarcinoma. A diagnostic challenge

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Dear Editor,

81-year-old woman. She is Ecuadorian, and has a history of high blood pressure and chronic ischemic heart disease. She was admitted in April 2021 due to acute cholangitis. CholangioRNM: bile duct dilation and possible obstruction in the distal common bile duct. Endoscopic-ultrasound: non-dilated common bile duct, distal asymmetric thickening of 15mm and biliary mud. ERCP: doubtful defect of suprapapillary repletion without extraction of material after sphincterotomy and balloon extractor passes.

A second episode of acute cholangitis occurred in Ecuador in December 2021. ERCP: hemobilia and radiolucent image in distal common bile duct without extraction of lithiasic material. It was brushed and a metal stent was inserted. Cytology: fibrinohematic material without malignancy. After five months, the patient developed a third acute cholangitis. ERCP: biliary stent obstruction due to lithiasis resolved with balloon extractor. Five months later, the patient had her fourth admission for cholangitis and confirmation of anemia. ERCP: spontaneous migration of biliary stent and hemobilia with clots. Direct-vision-cholangioscopy (SpyGlass®): unstable bleeding clot. Urgent CT-angiography: no collateral circulation in the hepatic or peribiliary hilum and no active bleeding. Conservative management was performed without hemorrhagic recurrence.

In January 2023 she was transferred to our center after developing her fifth acute cholangitis with an episode of hematemesis and hypotension. Urgent CT-angiography: no active bleeding and hyperdense contents in the bile duct. ERCP: dilated extrahepatic bile duct with repletion defects (Figure 1a). The Fogarty-balloon was introduced, washed with serum and the clots were extracted. SpyGlass®-cholangioscopy: neoproliferative lesion in distal common bile duct, 15mm from the papilla (Figure 1b), suggestive of cholangiocarcinoma. Biopsies compatible with biliary adenocarcinoma. Extension study without distance disease. Under a multidisciplinary approach, cephalic duodenopancreatectomy was decided. Study of surgical sample: pT2pN2 (Figure 1c-d).

The etiology of hemobilia has mainly iatrogenic (>50%), followed by traumatic causes. Others are biliopathy due to portal high pressure, or neoplastic or infective^{1,2,3,5} biliopathy. In the case of non-clear hemobilia, direct-vision-cholangioscopy can change the management in >34% of cases⁴. Our patient had episodes of obstructive hemobilia with secondary cholangitis without objectifying underlying pathology. When she was referred to our center, SpyGlass®-cholangioscopy identified the suspicious lesion compatible with early-stage cholangiocarcinoma despite the diagnostic delay. In

conclusion, it is important to keep in mind the neoformative etiology as a potential cause of hemobilia of unclear origin, in which case, cholangioscopy (SpyGlass®) can contribute to the recognition of the signs of malignancy of the lesion and, therefore, to the diagnosis.

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FIGURE 1:

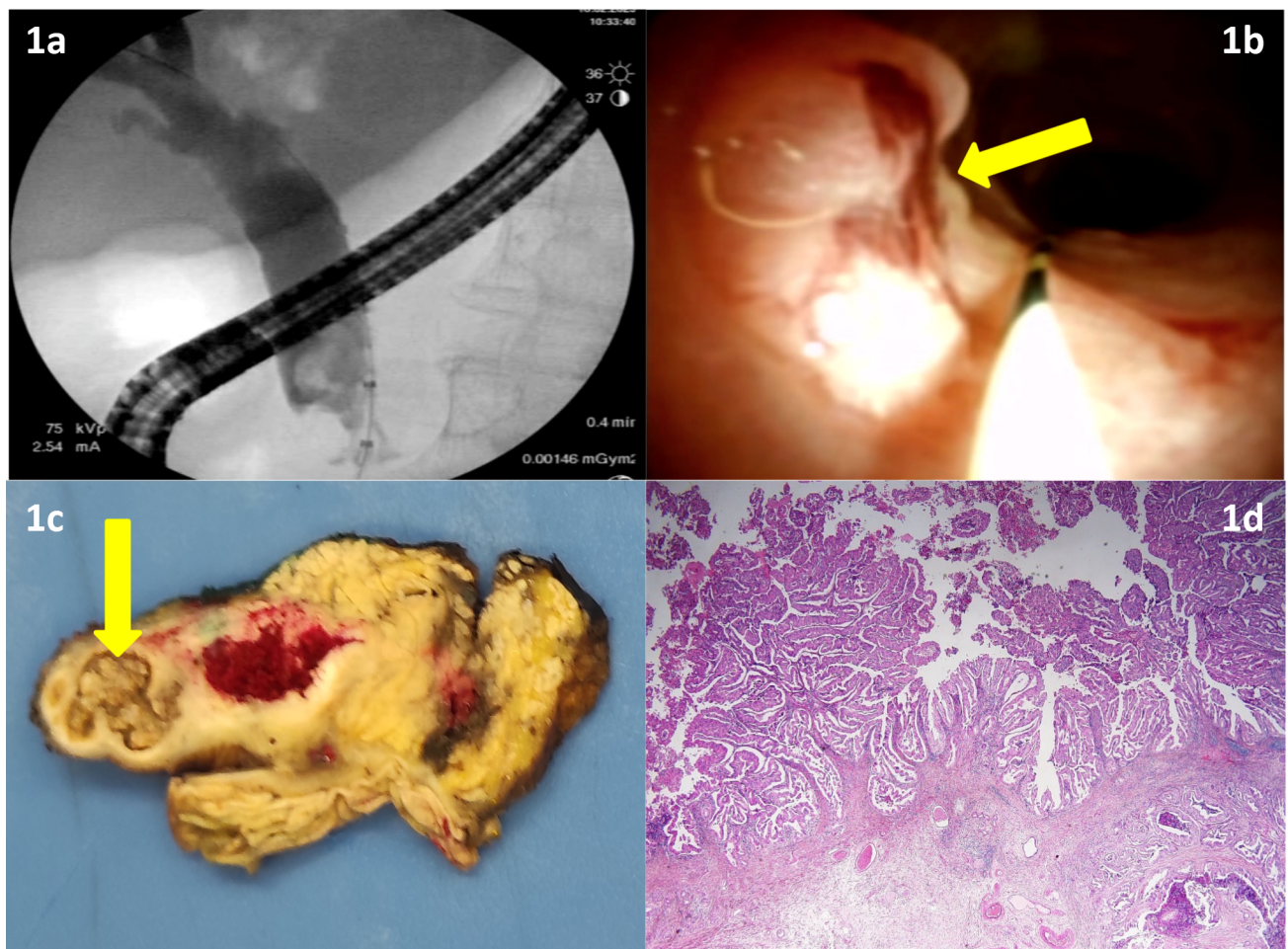


Figure 1: Recurrent hemobilia secondary to distal cholangiocarcinoma. 1a: Moderately dilated extrahepatic bile duct with several repletion defects in its distal portion. 1b: Neoproliferative and friable lesion in distal common bile duct (yellow arrow). 1c: Cephalic duodenopancreatectomy surgical sample where cholangiocarcinoma is objectified (yellow arrow). 1d: biliary adenocarcinoma.